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Adenomyoma of the distal common bile duct mimicking cholangiocarcinoma: A case report and narrative review

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Introduction

Adenomyomatosis is an uncommon benign proliferative lesion within the gastrointestinal tract. It most frequently involves the gallbladder fundus, but rare cases have also been reported in the stomach, small intestine, ampullary region, and extrahepatic biliary tree. Involvement of the common bile duct (CBD) is exceedingly rare and represents a significant diagnostic challenge because the lesion is symptomatic and often mimics malignant biliary strictures, particularly cholangiocarcinoma, both clinically and radiologically^[1-2]. The widespread use of advanced imaging modalities such as ultrasonography (USG), computed tomography (CT), magnetic resonance cholangiopancreatography (MRCP), and endoscopic retrograde cholangiopancreatography (ERCP) has improved the detection of biliary lesions. However, distinguishing benign adenomyomatous lesions from early cholangiocarcinoma remains difficult because there are no pathognomonic radiological features. Patients commonly present with obstructive jaundice, abdominal pain, pruritus, or biliary obstruction, further increasing suspicion for malignancy. Consequently, most cases are diagnosed only after radical surgical excision and subsequent histopathological examination^[1-6]. Herein, we report a rare case of adenomyomatous hyperplasia (AH) involving the distal CBD that clinically, radiologically, and cytologically mimicked distal cholangiocarcinoma, resulting in

pancreaticoduodenectomy. This case highlights the diagnostic dilemma associated with this entity and emphasizes the need for improved preoperative diagnostic approaches to avoid unnecessary extensive surgical interventions.

The study was granted an exemption from ethical review by the Institutional Review Board of Tata Main Hospital. Written informed consent was obtained from the patient.

Case report

A 69-year-old male presented with a 10-day history of fever and abdominal pain. He had a history of paranoid schizophrenia and hypertension and had no significant history of weight loss or anorexia. On physical examination, the patient had icterus and pruritus suggestive of obstructive jaundice. No palpable abdominal mass or organomegaly was identified. Laboratory investigations revealed serum lipase was normal (54.6 U/L), whereas serum amylase was mildly elevated (161.9 U/L). Fluid amylase (180.7 U/L) and fluid lipase (231.8 U/L) were also elevated, whereas total protein levels were reduced. Tumor markers including carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA19-9) were within normal limits. Ultrasonography and CT scan (**Fig. 1**) demonstrated bilobar intrahepatic biliary radicle dilatation with dilatation of the CBD. MRCP revealed a distal CBD stricture associated with proximal biliary dilatation, raising a strong suspicion for distal cholangiocarcinoma (**Fig. 2**). Brush

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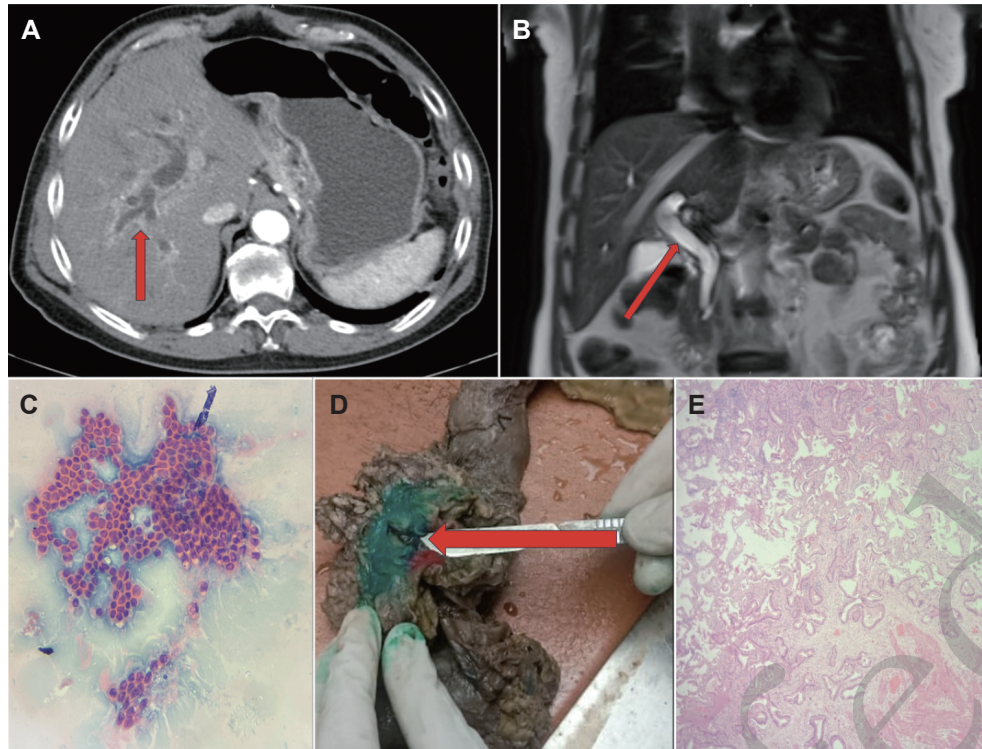


Fig. 1 Multimodal evaluation of a common bile duct (CBD) lesion mimicking biliary Malignancy. A: CT scan showing dilated CBD and intrahepatic biliary radicles (red arrow). B: Dilated CBD with abrupt narrowing in MRCP (red arrow). C: Cytology showing sheets of epithelial cells with cytological atypia, suggesting high-grade biliary intraepithelial neoplasia (Category IVb) (PAP stain, 200×). D: Gross photograph showing obstruction of the CBD by a well-circumscribed nodular lesion (red arrow). E: Proliferated glands with surrounding smooth muscle (hematoxylin and eosin stain, 200×). Abbreviations: CT, computed tomography scan; CBD, common bile duct; MRCP, magnetic resonance cholangiopancreatography; PAP stain: Papanicolaou stain.

cytology showed moderately cellular smears composed of three-dimensional clusters, sheets, and singly scattered epithelial cells with occasional acinar formations. The cells exhibited hyperchromatic nuclei, coarse chromatin, scant to moderate cytoplasm, irregular nuclear contours, nuclear overlapping, and increased nuclear-to-cytoplasmic ratio in focal areas. Mild anisonucleosis was also noted. The background was hemorrhagic with mucoid material. Based on these findings, a cytological diagnosis consistent with high-grade biliary intraepithelial neoplasia (BilIN) (Category IVb) was rendered ([Fig. 3](#)).

The case was discussed in a multidisciplinary tumor board meeting. After an informed discussion with the patient and family members, pancreaticoduodenectomy was performed. The resected specimen included the distal stomach, proximal duodenum, part of the pancreas, gallbladder, distal CBD, and lymph nodes from stations 8 and 13. The postoperative course was uneventful, and the patient was discharged on postoperative day 13 in stable condition.

Gross examination revealed a dilated CBD with a firm, well-circumscribed nodular lesion measuring $0.7 \times 0.7 \times 0.6$ cm located at the distal end of the duct,

causing luminal narrowing and obstruction ([Fig. 4](#)). The remaining pancreas, stomach, duodenum, and gallbladder appeared grossly unremarkable. Microscopic examination demonstrated a nodular lesion confined to the distal CBD wall. The lesion was composed of proliferated glandular structures lined by uniform cuboidal to columnar epithelium embedded within prominent bundles of smooth muscle fibers. The glands extended into and beyond the muscularis mucosa ([Fig. 5](#)). However, there was no evidence of cytological atypia, significant mitotic activity, desmoplastic stromal response, infiltrative growth pattern, lymphovascular invasion, or perineural invasion. The surrounding mucosa showed chronic inflammatory changes without dysplasia. All resection margins and sampled lymph nodes were negative for malignancy. The main histological differential diagnosis was well-differentiated adenocarcinoma. Immunohistochemistry was therefore performed to support the morphological diagnosis. CK19 highlighted the biliary epithelial component, while desmin confirmed the smooth muscle bundles surrounding the glandular structures. No features suggestive of malignancy were identified. Therefore, a final diagnosis of adenomyoma of the distal CBD was

established. The patient has remained asymptomatic during a 2-year follow-up period. Liver function tests, including serum bilirubin, aspartate aminotransferase (AST), alanine aminotransferase (ALT), alkaline phosphatase (ALP), and serum calcium levels, were within normal limits. Follow-up ultrasonography showed no evidence of recurrence or residual lesion.

Discussion

Adenomyomatosis is a benign proliferative lesion characterized by epithelial and smooth muscle hyperplasia with formation of intramural diverticula known as Rokitsky–Aschoff sinuses. Morphologically, it may occur in focal, annular, or diffuse forms. The first case involving the hepatic bile duct was described by Dowdy *et al.* in 1962^[3]. The exact pathogenesis of AH remains uncertain. Most authors consider it a hamartomatous or hyperplastic lesion. One widely accepted theory suggests that these lesions may represent an incomplete form of heterotopic pancreas, particularly type III heterotopia described by von Heinrich in 1909. Chronic inflammation and increased intraluminal pressure may also contribute to epithelial proliferation and smooth muscle hypertrophy^[1,6].

An extensive review of the literature in PubMed and Google Scholar, using the MeSH terms adenomyoma or adenomyomatous hyperplasia of CBD or extrahepatic location, revealed approximately 60 reported cases of extrahepatic adenomyomatosis involving the cystic duct, common hepatic duct, right and left hepatic ducts, ampulla of Vater, and CBD. However, isolated adenomyoma confined to the CBD is particularly rare, with only about 23 cases reported in the published literature, as shown in [Table I](#)^[1,3–10,15–27]. Lesions have been described in proximal, middle, and distal portions of the CBD, including one case associated with a choledochal cyst^[4]. Including the present patient, 25 cases were available for analysis. Seventeen of the 25 reported patients (68%) underwent extensive surgery, including Whipple procedure, pancreaticoduodenectomy, distal pancreaticoduodenectomy, and pylorus-preserving pancreaticoduodenectomy. Eight cases (32%) underwent limited or local procedures, including local excision of the lesion, CBD resection with biliary reconstruction, cholecystectomy, endoscopic sphincterotomy, endoscopic excision, and choledochal cyst excision with hepaticojejunostomy. The predominance of extensive surgeries highlights the major diagnostic challenge posed by CBD adenomyoma. In most cases, the lesion clinically and

radiologically mimicked distal cholangiocarcinoma, leading surgeons to perform radical resections to exclude malignancy definitively. Only a minority of patients underwent conservative or localized procedures, usually when intraoperative frozen section evaluation or less suspicious imaging findings allowed exclusion of invasive carcinoma.

Radiological evaluation plays an important role in localizing biliary obstruction but lacks specificity in distinguishing benign from malignant strictures. Ultrasonography is generally the first-line imaging modality and helps identify biliary dilatation. MRCP provides excellent visualisation of the biliary tree and extent of obstruction, while CT scan is useful for evaluating suspected malignancy and surrounding structures. ERCP permits direct visualization, therapeutic intervention, and tissue sampling. Nevertheless, none of these modalities demonstrates characteristic findings specific to adenomyoma^[1,5,9–10]. Radiologically, adenomyoma of the CBD should be differentiated from well-differentiated cholangiocarcinoma. Adenomyoma typically appears as a short-segment, smooth, symmetric mural thickening or well-defined stricture with proximal biliary dilatation, without periductal infiltration, vascular encasement, or significant lymphadenopathy. In contrast, cholangiocarcinoma usually demonstrates irregular or asymmetric wall thickening, progressive contrast enhancement, periductal extension, vascular invasion, and regional lymph node enlargement. Unlike gallbladder adenomyomatosis, CBD adenomyoma lacks characteristic imaging signs such as the comet-tail artifact and pearl-necklace sign, making definitive differentiation by imaging alone difficult and necessitating histopathological confirmation^[11–13].

Brush cytology obtained during ERCP is commonly used because it is minimally invasive and technically feasible. However, the sensitivity of brush cytology for malignant biliary strictures varies widely from 24.6% to 83%, although specificity remains high at 91.7%–100%. False-negative results may occur due to inadequate sampling, submucosal location of lesions, technical limitations, or well-differentiated tumors with bland cytology. Conversely, false-positive interpretations may arise in inflammatory and reactive conditions such as primary sclerosing cholangitis, IgG4-related cholangitis, chronic pancreatitis, or epithelial reactive atypia^[9].

In the present case, brush cytology suggested high-grade BILIN, leading to a strong suspicion of malignancy as shown in [Fig. 3](#). This likely represented a false-positive interpretation due to

Reference	Age/Sex	Pre-operative findings	Management
Gilbert LA <i>et al.</i> (1985) ^[17]	70/M	Distal CBD	Cholecystectomy
Cook DJ <i>et al.</i> (1988) ^[18]	82/F	Proximal CBD	Transduodenal Sphincteroplasty with cholecystectomy
Ikei <i>et al.</i> (1989) ^[19]	71/F	Dilated CBD and stenosis of distal part in PTC	Whipple procedure
Ikei <i>et al.</i> (1989) ^[19]	52/F	Dilated CBD with mural irregularity and spherical immobile shadow defect in ERCP	Whipple procedure
Legakis <i>et al.</i> (1990) ^[20]	55/F	Not done	Choledochal excision and hepatojejunal anastomosis
Thomas <i>et al.</i> (1990) ^[21]	68/F	Not done	Whipple procedure
Matsumoto <i>et al.</i> (1992) ^[22]	46/M	CT showed enhanced thickening of distal CBD	Pancreaticoduodenectomy
Sudan N, Lafon J <i>et al.</i> (1992) ^[23]	42/M	Distal CBD	Pancreaticoduodenectomy
Läuffer JM <i>et al.</i> (1998) ^[3]	69/F	Hemispherical contrast defect in terminal CBD, 1.5 cm proximal to ampulla	Frozen section evaluation followed by a local excision of the tumor in the distal CBD through the papilla of Vater
Tsukamoto <i>et al.</i> (1999) ^[7]	31/F	CBD stenosis	Resection of the CBD with the tumor was performed with choledochojejunostomy reconstruction.
Singh <i>et al.</i> (2000) ^[4]	45/F	Distal CBD nodule along with choledochal cyst	Excision of choledochal cyst with Roux-en-Y hepaticojejunostomy was performed. A segment of dilated common bile duct and an attached nodule were received.
Ojima <i>et al.</i> (2000) ^[8]	64/F	Possible stenosis in distal CBD in ERCP	Distal pancreaticoduodenectomy
Jang <i>et al.</i> (2005) ^[24]	52/F	Distal CBD polypoidal lesion with diffuse dilatation of the CBD	Pylorus-preserving pancreaticoduodenectomy
Iwaki <i>et al.</i> (2008) ^[5]	62/F	Thickening of lower CBD wall in CT with stenosis of lower CBD in ERCP	Pylorus-preserving pancreaticoduodenectomy was performed
Shu <i>et al.</i> (2008) ^[25]	51/M	Dilatation and irregular stricture of CBD with mass in the pancreatic segment of CBD in MRCP	Distal pancreaticoduodenectomy
Genevay <i>et al.</i> (2009) ^[15]	73/M	CBD dilatation with stenosis in distal part, 1cm above the ampulla	Distal pancreaticoduodenectomy
Choi <i>et al.</i> (2016) ^[26]	42/M	Abrupt narrowing of the distal CBD with proximal dilatation in CT	Distal pancreaticoduodenectomy
D'Assuncao <i>et al.</i> (2016) ^[27]	50/F	Dilated CBD in MRI, EUS showed a hypoechoic lesion near the papilla	Endoscopic excision using biopsy forceps
Chandler <i>et al.</i> (2018) ^[6]	63/M	Distal CBD dilatation in Ultrasound	Whipple procedure
Xu <i>et al.</i> (2019) ^[16]	68/F	Proximal CBD dilatation in MRCP and distal obstruction in Ultrasound	Local resection
Gouveia <i>et al.</i> (2021) ^[9]	70/F	CBD dilatation and localized stenosis in MRCP	Whipple procedure
Tan <i>et al.</i> (2022) ^[10]	74/M	Calculus in distal CBD in CT	Cholecystectomy with open CBD resection
Weng <i>et al.</i> (2025) ^[11]	54/F	Significant dilation of the CBD with a contrast-enhanced tumor-like mass measuring approximately 1.6 cm in the distal CBD,	Whipple procedure
Present case, 2026	69/M	ERCP revealed a distal CBD stricture with proximal biliary dilatation	Whipple procedure

Abbreviations: CBD, common bile duct; PTC, percutaneous transhepatic cholangiography; ERCP, endoscopic retrograde cholangiopancreatography; CT, computed tomography scan; MRCP, magnetic resonance cholangiopancreatography.

reactive atypia and overlapping cytomorphological features. The cytological and architectural patterns can lead to false positivity. For example, exaggerated downward invagination and reactive mucosal

crowding can produce nuclear enlargement and overlap resembling high-grade dysplasia. Interlacing smooth muscle fibers and fibroblasts create a firm, desmoplastic-like environment that falsely suggests an

invasive malignant process. Additionally, disoriented hyperplastic glands sampled *via* fine-needle aspiration lack smooth polarity, which cytopathologists can misinterpret as preinvasive or *in situ* high-grade BILIN. Such diagnostic uncertainty often results in radical surgical procedures, including pancreaticoduodenectomy or bile duct excision, to definitively exclude malignancy^[1,3,11].

Hence, the diagnostic yield of biliary sampling may improve when multiple modalities are combined, including brush cytology, intraductal biopsy, and endoscopic ultrasound-guided fine-needle aspiration (EUS-FNA). Rapid on-site evaluation (ROSE) and biopsy ROSE (B-ROSE) or imprint smears from biopsies can further enhance adequacy assessment, optimizing the number of needle passes performed and improving diagnostic accuracy during procedures^[14]. Endoscopic ultrasound-guided fine-needle biopsy (EUS-FNB), particularly with newer end-cutting designs, has been shown to preserve the cellular architecture and has become an increasingly useful tool in establishing a definitive diagnosis of malignancy in a variety of solid lesions, including pancreatic tumors^[12]. The efficacy of EUS-FNA with ROSE was similar to that of EUS-FNB without ROSE, even with newer end-cutting needles. Frozen section evaluation may also assist intraoperative decision-making and help determine the extent of surgery^[14].

Histopathologically, adenomyoma is characterized by benign glandular proliferation within hypertrophied smooth muscle stroma without atypia, mitotic activity, desmoplasia, or invasive growth. Although rare cases may show limited perineural involvement, true malignant features are absent, and serosal invasion is not seen. Immunohistochemistry can be useful in excluding well-differentiated adenocarcinoma, particularly in limited or challenging specimens^[1-2]. Complete surgical excision is considered curative, and recurrence has not been documented in the available literature, although follow-up is limited in many reports. This case highlights the considerable diagnostic challenges associated with CBD adenomyoma and emphasizes the importance of integrating clinical, radiological, cytological, and histopathological findings before undertaking radical surgery whenever feasible.

Conclusions

AH of the CBD is an exceptionally rare benign lesion that closely mimics cholangiocarcinoma clinically, radiologically, and even cytologically.

Because imaging findings are nonspecific and biliary brush cytology has limited sensitivity and occasional false-positive interpretations, definitive preoperative diagnosis remains difficult. As a result, many patients undergo extensive surgical procedures to exclude malignancy. The present case highlights the importance of considering adenomyoma in the differential diagnosis of distal CBD strictures, particularly in patients with normal tumor markers and inconclusive investigations. Histopathology remains the gold standard for diagnosis, while immunohistochemistry may aid in differentiating the lesion from well-differentiated adenocarcinoma. Increased awareness of this rare entity among clinicians, surgeons, radiologists, and pathologists is essential to avoid overtreatment and unnecessary radical surgery. Future advances in multimodal tissue sampling and minimally invasive diagnostic techniques may improve preoperative diagnostic accuracy and facilitate more conservative management strategies in selected patients.

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Author contributions

Sushma Bharti and Sapna Sandeep Saha: Study concept, Study design, Data collection & processing, Analysis and interpretation, Literature search, Writing. **Neetesh Kumar Sinha, Farha Rana, and Radhika Narayana:** Study concept, Analysis and interpretation, Literature search, Writing.

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